

# Midline (Dermoid) Cysts of the Floor of the Mouth: Report of 16 Cases and Review of Surgical Techniques

Francesco Longo, M.D., Ph.D., Pietro Marenmonti, M.D., Ph.D., Giuseppe Michele Mangone, M.D., Giuseppe De Maria, M.D., and Luigi Califano, M.D.

*Naples and Bologna, Italy*

A retrospective review of 16 cases of midline (dermoid) cysts of the floor of the mouth is presented, evaluating the different surgical approaches. Sixteen cases of patients with a diagnosis of midline cyst of the floor of the mouth, treated at the Maxillofacial Surgery Department of the School of Medicine and Surgery of the “Federico II” University of Naples (Naples, Italy), were observed over a 10-year period, between 1988 and 1998; age, sex, localization, diagnostic technique, and type of treatment were evaluated. Male patients were more frequently affected, with a male-to-female ratio of 3:1 (12:4 cases). Patients ranged in age from 5 to 51 years (average age, 27.8 years). The preoperative assessment was made using ultrasonography in all cases but one, computed tomography in eight cases, and magnetic resonance imaging in three cases. Regarding surgical techniques used, a transcutaneous approach was adopted for median geniohyoid cysts, an extended median glossotomy technique was used for very large median genioglossal cysts, a median glossotomy technique was used for median genioglossal cysts, and a midline incision of the oral mucosa along the lingual frenulum was used for sublingual cysts. During the postoperative course, there were no complications except for modest edema in three cases. Follow-up ranged between 24 months and 12 years; no relapses or malignant changes were observed. In the authors’ experience, the intraoral approach was also effective for the treatment of large lesions and led to very good cosmetic and functional results, whereas the extraoral incision was necessary only when the cysts were under the geniohyoid muscle. (*Plast. Reconstr. Surg.* 112: 1560, 2003.)

Midline (dermoid) cysts of the floor of the mouth are quite uncommon. In the classic article by New and Erich,<sup>1</sup> only 1.6 percent (24 cases) of 1495 described cases of dermoid cysts were located in the floor of the mouth; their

low incidence is confirmed in other recent publications.<sup>2-8</sup> In fact, dermoid cysts occur primarily in the testes and ovaries, and the most common location in the head and neck is the external third of the eyebrow.<sup>5</sup>

The pathogenesis of midline cysts of the floor of the mouth is not well established, and dysontogenetic, traumatic, and thyroglossal anomaly theories have been suggested. Histologically, Meyer divided the cysts of the floor of the mouth into three groups: epidermoid, dermoid, and teratoid. Although dermoid cysts represent a separate entity, the term “dermoid” is typically used to indicate all three categories.<sup>5</sup>

Anatomically, it is possible to distinguish three different types of dermoid cysts: median genioglossal, median geniohyoid, and lateral, according to anatomic relationship between the cyst and the muscles of the floor of the mouth. Lateral cysts are very rare, and some authors consider them to be median cysts that have laterally expanded and not a separate entity.

Dermoid cysts generally present slow and progressive growth, and even if they are congenital, the diagnosis is commonly possible in the second or third decade of life.<sup>8</sup> The treatment of dermoid cysts of the floor of the mouth is surgical and can be intraoral or extraoral according to the localization and the size of the mass. In this article, we present a

From the Department of Ear, Nose, and Throat–Head and Neck Surgery, National Cancer Institute “Fondazione Pascale”; Department of Maxillofacial Surgery, Bellaria Hospital; and Department of Maxillofacial Surgery, School of Medicine and Surgery “Federico II,” University of Naples. Received for publication September 18, 2002; revised January 27, 2003.

DOI: 10.1097/01.PRS.0000086735.56187.22

retrospective review of 16 cases treated with different surgical approaches to evaluate the outcomes of various treatment options and to propose a standardization of terminology and surgical procedures.

#### PATIENTS AND METHODS

We retrospectively examined the cases of 16 patients diagnosed as having midline dermoid cyst of the floor of the mouth and treated at the Maxillofacial Surgery Department of the School of Medicine and Surgery of the "Federico II" University of Naples (Naples, Italy). The patients were observed over a 10-year period between 1988 and 1998. The parameters considered in this study were age, sex, localization, diagnostic technique, and surgical approach.

The preoperative assessment consisted of ultrasonography (Fig. 1, *left*) and/or computed tomography (Fig. 1, *right*) and/or magnetic resonance imaging. A fine-needle aspiration biopsy was performed in all cases.

Regarding the surgical procedure, the following approaches were adopted: a transcutaneous approach for cases of median geniohyoid cysts (Fig. 2, *left*), an extended median glossotomy technique (eventually associated with partial evacuation) for cases of very large median genioglossal cysts (Fig. 2, *right*), a median glossotomy technique for cases of median genioglossal cysts (Fig. 3, *left*), and a midline incision of the oral mucosa along the lingual frenulum for cases of sublingual cysts. The operations were performed with the patient under general anesthesia with rhinotracheal intu-

bation, except in the event of extraoral incision. An aspirative drain was placed and generally removed after 2 days, and an antibiotic (cephalosporin intramuscularly) was administered to the patient preoperatively and for 2 days postoperatively. Patients were followed up for 2 years or more, with clinical examination and ultrasonography twice a year.

#### RESULTS

Our findings are summarized in Table I. Patients ranged in age from 5 to 51 years (average age, 27.8 years). Male patients were affected more frequently, with a male-to-female ratio of 3:1 (12:4 cases).

The preoperative assessment was made using ultrasonography in all cases but one, computed tomography in eight cases, and magnetic resonance imaging in three cases. The fine-needle aspiration biopsy showed the features of epidermoid cyst in 14 cases and epidermoid cyst in two cases; the microscopic examination of the specimens confirmed the preoperative diagnosis in all cases. The cysts were located above the geniohyoid muscles in 10 cases and in the submental region between the geniohyoid and the mylohyoid muscles in six cases.

A median glossotomy was performed in two cases, an extended median glossotomy with ventral incision was performed in six cases, a median mucosal incision was performed in two cases, and a submental access was performed in six cases. Anesthesia was generally easily administered, except for one patient, who needed endoscopic intubation.

During the postoperative course, there were

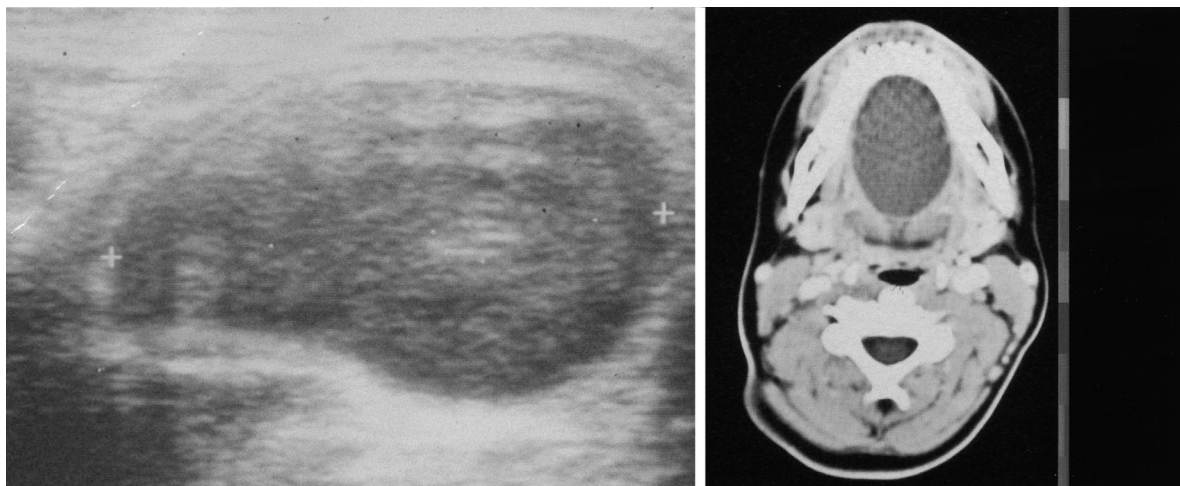


FIG. 1. Diagnostic techniques. (*Left*) Ultrasound of a dermoid cyst of the floor of the mouth. (*Right*) Computed tomographic scan of a dermoid cyst of the floor of the mouth.

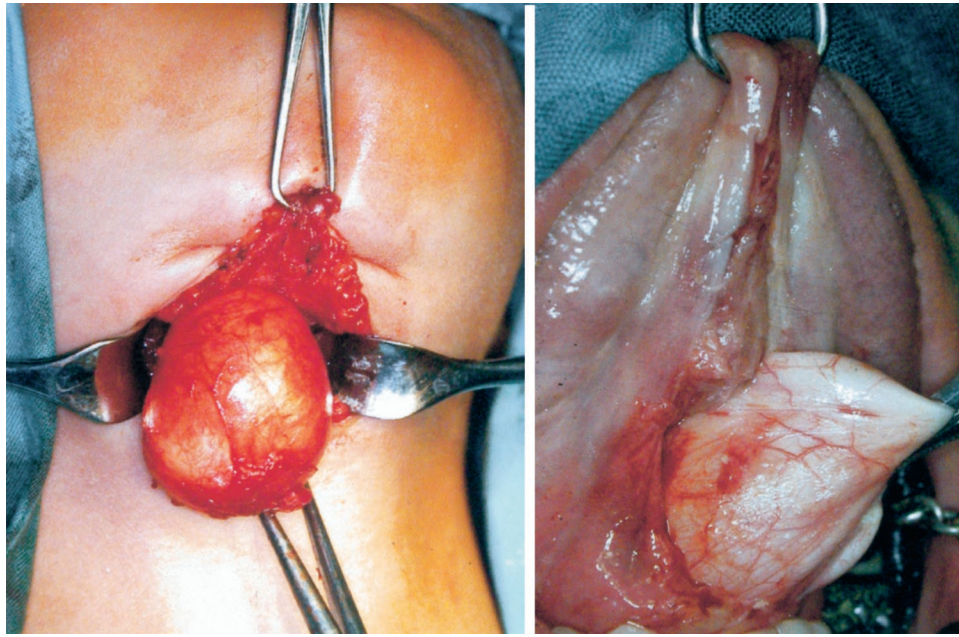


FIG. 2. Surgical techniques. (Left) Transcutaneous approach. (Right) Extended median glosotomy approach.

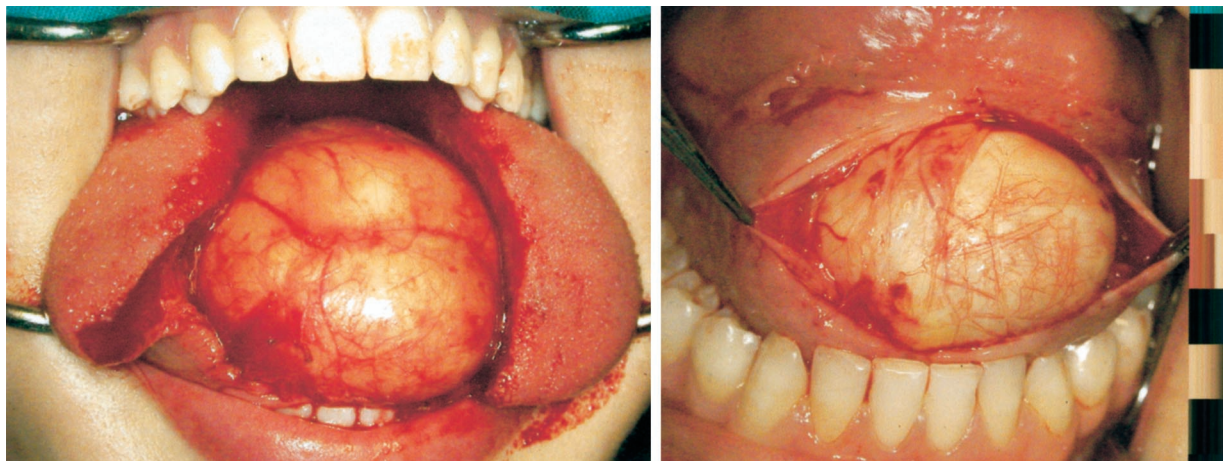


FIG. 3. Surgical techniques. (Left) Median glosotomy approach. (Right) Midline incision of the floor of the mouth.

no complications except for a modest edema in three cases. Follow-up ranged between 24 months and 12 years (average, 6 years); no relapses or malignant changes were observed.

#### DISCUSSION

Midline dermoid cysts of the floor of the mouth are painless lesions that swell from the anterior portion of this region. Because they can displace the tongue, patients usually present with dysphagia, dysphonia, and dyspnea, and in the case of lower localization, they present a characteristic double chin.<sup>4-6</sup> Dermoid cysts are generally diagnosed in young adults in the second and third decades of life;

however, we did find one case in a 5-year-old patient; no sex correlation is demonstrated,<sup>4</sup> although Howell does report a higher frequency in male patients.<sup>5</sup> There are no rules regarding the timing for the operation; because dermoid cysts are mainly congenital, they can appear in every age of life, so when they appear (generally with dysphagia, dysphonia, and dyspnea), it is generally the right time to operate on them. Also, in very young patients, a problem can arise from the anesthesiologic risk, which is generally quite low in patients weighing more than 20 kg.

Dermoid cysts may be classified as congenital or acquired, even if there is no difference be-

TABLE I  
Patient Summary and Features of the Lesions

Patient	Sex	Age (yr)	Imaging	FNAB	Histology	Anatomy	Dimension (cm)	Treatment
1	M	51	US, CT, MRI	Epidermoid cyst	Epidermoid cyst	Median geniohyoid	8 × 5	SA
2	M	28	US	Epidermoid cyst	Epidermoid cyst	Median genioglossal	6.5 × 4.5	EMG
3	F	15	CT	Epidermoid cyst	Epidermoid cyst	Median genioglossal	6 × 5	MG
4	M	5	US, CT	Epidermoid cyst	Epidermoid cyst	Median geniohyoid	5 × 3	SA
5	M	17	US	Epidermoid cyst	Epidermoid cyst	Median genioglossal	6.5 × 5	EMG
6	M	35	US, CT	Epidermoid cyst	Epidermoid cyst	Median geniohyoid	5.5 × 3.4	SA
7	F	24	US, CT	Dermoid cyst	Dermoid cyst	Median genioglossal	4.5 × 2.5	MMI
8	M	31	US	Epidermoid cyst	Epidermoid cyst	Median genioglossal	5.5 × 4	EMG
9	M	39	US, MRI	Epidermoid cyst	Epidermoid cyst	Median genioglossal	6.7 × 4.5	MG
10	M	19	US	Epidermoid cyst	Epidermoid cyst	Median genioglossal	7.5 × 6.7	EMG
11	F	27	US, CT	Epidermoid cyst	Epidermoid cyst	Median genioglossal	6.5 × 5.4	EMG
12	M	44	US	Dermoid cyst	Dermoid cyst	Median geniohyoid	5.5 × 4.3	SA
13	M	37	US, CT	Epidermoid cyst	Epidermoid cyst	Median geniohyoid	6.5 × 4.5	SA
14	F	32	US	Epidermoid cyst	Epidermoid cyst	Median genioglossal	4 × 2.5	MMI
15	M	21	US, MRI	Epidermoid cyst	Epidermoid cyst	Median genioglossal	6 × 4.5	EMG
16	M	30	US, CT	Epidermoid cyst	Epidermoid cyst	Median geniohyoid	5 × 3.5	SA

FNAB, fine-needle aspiration biopsy; CT, computed tomography; MRI, magnetic resonance imaging; US, ultrasonography; MG, median glossectomy; EMG, extended median glossectomy; MMI, median mucosal incision; SA, submental access.

tween the two on presentation or histologically. Congenital cysts are dysembryogenetic lesions that arise from ectodermic elements entrapped during the midline fusion of the first and second branchial arches between the third and fourth weeks of intrauterine life<sup>6-8</sup>; alternatively, they may arise from the tuberculum impar of His which, with each mandibular arch, forms the floor of the mouth and the body of the tongue.<sup>5</sup> Acquired cysts derive from traumatic or iatrogenic inclusion of epithelial cells or from the occlusion of a sebaceous gland duct.<sup>6,9-11</sup> Moreover, others authors proposed that midline cysts may represent a variant form of thyroglossal duct cyst.<sup>10</sup>

Histologically, midline dermoid cysts of the floor of the mouth are classified according to Meyer's classification,<sup>7</sup> thus dividing them into three groups: epidermoid cysts, which consist of an epithelial-lined wall that may be partly keratinized; dermoid cysts, which are epidermoid-like cysts but show evidence of skin appendages, such as hair follicles, hair, sweat, and sebaceous glands; and teratomas, which contain, in addition to skin appendages, mesodermal elements such as bone, muscle, respiratory and gastrointestinal tissues, and a fibrous capsule. The latter type is the only variety that may have a malignant change.<sup>12</sup>

Anatomic classification divides the cysts of the floor of the mouth into three groups according to their relation to the muscles of the floor of the mouth<sup>13</sup>: sublingual or median genioglossal cysts, located above the geniohyoid muscles; median geniohyoid cysts, located

in the submental region between the geniohyoid and mylohyoid muscles; and lateral cysts, located in the submaxillary region.

Many other lesions, such as an infectious process, ranulas, or benign neoplasm of lipoid, salivary, neural, or vascular tissues, may involve the floor of the mouth, as they present a similar clinical appearance. For this reason, bimanual palpation and conventional radiography are not always sufficient in making differential diagnoses. In these cases, it is necessary to use ultrasonography, computed tomography, or magnetic resonance imaging together with cytologic examination by fine-needle aspiration biopsy.<sup>10,11</sup> All of our patients except one were studied using bimanual palpation, ultrasonography (Fig. 1, *left*), and fine-needle aspiration biopsy. In some cases, computed tomography (Fig. 1, *right*) or magnetic resonance imaging was also performed. Ultrasonography represents the first choice of imaging technique because it is reliable, economical, and without x-ray exposure, so it is easily suitable for young patients also. Computed tomography and magnetic resonance imaging allow more precise localization of the lesion in relationship to geniohyoid and mylohyoid muscles, and they also enable the surgeon to choose the most appropriate surgical approach, especially for very large lesions. The cytologic examination is the only way to achieve a sure preoperative diagnosis; therefore, in our opinion, it is necessary to perform a fine-needle aspiration biopsy to complete the preoperative assessment. A fine-needle aspiration biopsy is also an appropriate diagnostic technique in the very rare case of

TABLE II  
Surgical Approach According to Site and Size

Site and Size	Surgical Approach
Median geniohyoid	Submental (extraoral) access
Small or superficial median genioglossal	Median mucosal incision
Median genioglossal	Median glossotomy
Very large median genioglossal	Extended median glossotomy with partial evacuation of the cyst and/or submental (extraoral) access

malignant changes that can occur in teratoid cysts.

Surgical enucleation is the only effective treatment for these kinds of lesions. Several techniques are reported in the literature, which may be divided into intraoral and extraoral techniques depending on which approach is used (Table II).

In the case of an intraoral approach, a midline vertical, mucosal incision is performed along the ventral surface of the tongue; however, only small cysts can be enucleated using this kind of incision.<sup>10</sup> Lowry et al.<sup>6</sup> describe a bilateral incision along the mandibular ridge crest, Brusati et al.<sup>11</sup> propose a midline glossotomy, and Di Francesco et al.<sup>14</sup> describe a modification of this surgical technique consisting of an extension of the incision along the ventral surface of the tongue associated with partial evacuation of the cyst. The latter two techniques, in our opinion, allow us to obtain a very good approach to the cyst and to obtain adequate surgical control of the lesion in the event of median cysts located above the geniohyoid muscles.

The transcutaneous approach consists of a submental incision and a sharp, blunt dissection to reach and enucleate the lesion.<sup>6</sup> McGregor<sup>15</sup> describes a symphyseal mandibular osteotomy to enucleate a very large sublingual dermoid cyst. The extraoral approach is generally preferred in the case of median geniohyoid or very large sublingual cysts, whereas the intraoral approach is typically used for smaller sublingual cysts.<sup>6</sup>

We adopted a transcutaneous approach in six cases of median geniohyoid cysts (Fig. 2, *left*), the extended median glossotomy technique in six cases of median genioglossal cysts (Fig. 2, *right*), the median glossotomy technique in two cases of median genioglossal cysts (Fig. 3, *left*), and a midline incision of the oral mucosa along the lingual frenulum in two cases of sublingual cysts according to lesion size and localization (Fig. 3, *right*). Anesthesia

generally did not present any difficulty, but in cases of a very large lesion, intubation can be a serious problem; in such cases, an endoscopic intubation may be necessary and should be planned, to avoid an emergency tracheotomy.

The postoperative course does not present any kind of problem because there is little alteration in function, edema is generally modest, and complications are unusual. In some cases, dermoid cysts of the floor of the mouth may become infected, with the formation of a sinus and intraoral or cutaneous fistulas.<sup>2</sup>

Prognosis is very good, with a very low incidence of relapse, usually related to bone remnant to the genial tubercles or to the hyoid bone. Malignant changes have been recorded in dermoid cysts by New and Erich<sup>1</sup> but not in the floor of the mouth, although a 5 percent rate of malignant transformation of oral dermoid cysts has been reported by other authors, but only for the teratoid type.<sup>8</sup>

#### CONCLUSIONS

We found that appropriate imaging techniques (i.e., ultrasonography, computed tomography, and magnetic resonance imaging), together with fine-needle aspiration biopsy, are very effective in preoperative diagnosis and in cases of malignant change, and are useful in surgical planning. In cases of sublingual cysts, in our experience, the intraoral approach is effective in the treatment of large lesions, as it leads to very good cosmetic and functional results. The extraoral incision is mandatory only when the cyst lies under the geniohyoid muscle.

Francesco Longo, M.D., Ph.D.  
Via Nicolardi, 97 (Parco Verde)  
I-80131 Napoli, Italy  
frlongo@hotmail.com

#### REFERENCES

1. New, G. B., and Erich, J. B. Dermoid cysts of head and neck. *Surg. Gynecol. Obstet.* 65: 48, 1937.

2. Seward, G. R. Dermoid cysts of the floor of the mouth. *Br. J. Oral Surg.* 3: 36, 1965.
3. Yoshimura, Y., Takada, K., Takada, M., Mimura, T., and Mozl, M. Congenital dermoid cyst of the sublingual region: Report of a case. *J. Oral Surg.* 28: 366, 1970.
4. Ferran, F., and Abifadel, M. Kyste dermoïde du plancher buccal: A propos d'un cas et revue de la littérature. *Ann. Chir. Plast. Esthet.* 35: 69, 1990.
5. Howell, C. J. T. The sublingual dermoid cyst: Report of five cases and review of the literature. *Oral Surg. Oral Med. Oral Pathol.* 59: 578, 1985.
6. Lowry, R. E., Tempero, R. M., and Davis, L. F. Epidermoid cyst of the floor of the mouth. *J. Oral Surg.* 37: 271, 1979.
7. Meyer, I. Dermoid cysts (dermoids) of the floor of the mouth. *Oral Surg. Oral Med. Oral Pathol.* 8: 1149, 1955.
8. Zachariades, N., and Skoura-Kafoussia, C. A life threatening epidermoid cyst of the floor of the mouth: Report of a case. *J. Oral Maxillofac. Surg.* 48: 400, 1990.
9. Erich, J. B. Sebaceous, mucous, dermoid and epidermoid cysts. *Am. J. Surg.* 50: 672, 1940.
10. Walstad, W. R., Solomon, J. M., Schow, S. R., and Ochs, M. W. Midline cystic lesion of the floor of the mouth. *J. Oral Maxillofac. Surg.* 56: 70, 1998.
11. Brusati, R., Galioto, S., Tullio, A., and Moscato, G. The midline sagittal glossotomy for treatment of dermoid cysts of the mouth floor. *J. Oral Maxillofac. Surg.* 49: 875, 1991.
12. Mosby, E. L., Robertson, G. R., and Sugg, W. E. Compound dermoid cyst of the floor of the mouth. *J. Oral Surg.* 32: 601, 1974.
13. Colp, R. Dermoid cyst of the floor of the mouth. *Surg. Gynecol. Obstet.* 40: 183, 1925.
14. Di Francesco, A., Chiapasco, M., Biglioli, F., and Ancona, D. Intraoral approach to large dermoid cysts of the floor of the mouth: A technical note. *Int. J. Oral Maxillofac. Surg.* 24: 233, 1995.
15. McGregor, I. A. Symphyseal mandibular osteotomy in the approach to sublingual dermoid cyst. *Br. J. Plast. Surg.* 44: 544, 1991.